Original Article

Evaluation of use of distraction osteogenesis in mandibular retrognathia and its effect on soft and hard tissues and airway

ABSTRACT

Objectives: Bone distraction is the process of new bone formation between the surfaces of bone segments gradually separated by incremental traction of soft tissues. These adaptive changes in the soft tissues allow for greater skeletal movements while minimizing the potential relapse. In this study, we are reporting our clinical experience with mandibular distraction used to achieve simultaneous skeletal and soft-tissue correction with distraction osteogenesis (DO).

Methodology: A total of five patients who reported to the department for the treatment of mandibular deficiencies were selected. Cephalometric studies were done preoperatively and postoperatively for hard tissue assessment. Predefined reference points were used for the clinical evaluation for the evaluation of soft tissues. Results were compared between preoperative and 1st-, 3rd-, 6th-, and 12th-month postoperatively. **Results:** The clinical observations in our study showed that there is a remarkable improvement in the facial esthetics. Cephalometric analysis has shown lengthening of the mandibular corpus and increase in the height of the vertical ramus. Certain minimal complications have also been noted.

Conclusion: Despite the few complications, DO has become a popular surgical modality due to its many advantages.

Keywords: Airway, complications, distraction histiogenesis, distraction osteogenesis, relapse

INTRODUCTION

Mandibular micrognathia and facial asymmetry are relatively uncommon abnormalities of the craniomaxillofacial complex. These are addressed via osteotomies followed by skeletal advancements and fixation, with or without interpositional bone grafts. Nevertheless, various limitations are associated with these treatment modalities. The limitations include inability to stretch the soft tissue envelope along with hard tissues during advancement and donor site morbidity when bone grafts are used. Thus, surgical intervention by osteotomies only permits acute changes in the spatial arrangement of bones with limited possibility of soft tissue adaptation resulting in relapse.

Distraction osteogenesis (DO) is the process of new bone formation between the surfaces of bone segments which are gradually separated by incremental traction. The technique of

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DO has been popularized by Ilizarov of Russia in early 1950s. However, it was Codivilla who first lengthened the femur

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in 1905 to correct limb length discrepancy. It was Snyder *et al.* who carried this technique to the Maxillofacial region in 1973 by performing it in a canine study. Later, McCarthy *et al.* successfully lengthened canine mandible and extensively studied the histological examinations which revealed a highly organized biologic process.^[1]

Distraction forces applied to the bone also create tension in the surrounding soft tissues, initiating a sequence of adaptive changes termed as "distraction histiogenesis."^[2] This overcomes the relapse as seen in traditional orthognathic procedures when acutely stretched in greater magnitude. The purpose of this study is to evaluate the hard and soft tissue changes along with pharyngeal airway associated with mandibular lengthening by distraction.

METHODOLOGY

A prospective study was carried at our teaching institute and hospital, after procuring clearance from the institutional ethical board. Patients with mandibular retrognathia and with severe airway obstruction were selected for this study. A total of five patients (2 males and 3 females) age ranging from 14 to 21 years were selected, the mean age being 17 years. Three of five patients had postankylotic mandibular micrognathia and remaining two had nonsyndromic mandibular micrognathia. The following were the inclusion criteria for the study:

Inclusion criteria

- 1. Patients with severe mandibular retrognathia
- 2. Radiographic evidence of mandibular deficiency >7 mm
- 3. Radiographic evidence of airway compromise
- 4. Medically fit patients for surgery.

The patients were subjected to a thorough evaluation of history, clinical, blood investigations, and radiographs. According to the survey protocol, photographs were taken preoperatively [Figure 1] and postoperatively [Figure 2]. Preoperative and postoperative orthopantomograms [Figure 3] were taken to assess the bone formation in the distracted site. Preoperative and postoperative lateral cephalograms [Figure 4] were taken to assess the mandibular advancements and the pharyngeal airway. The clinical measurements and the radiographic tracings done preoperatively were compared with 3rd-, 6th-, and 12th-month postoperatively, and the technique was assessed.

The clinical measurements were done between the fixed points [Figure 5].^[3]

- Medial commissure to the buccal commissure (BC)
- Lateral commissure (LC) to the BC

- LC to the external auditory meatus (EAC)
- BC to the EAC.

Lateral cephalometric studies were performed to assess the dimensions of the mandibular body and ascending ramus. Osseous changes which were produced postoperatively were compared to that of preoperative values using Burstone's analysis.^[4] The ramus height is measured from Porion (Po) to Gonion (Go), and the anteroposterior length of the body of the mandible is measured from Gonion (Go) to Menton (Me) [Figure 6]. Porion was used as a reference point for the ramus height measurement because some of the patients were treated by temporomandibular joint ankylosis and lacked Condylion point. The upper airway is measured from base of the tongue to the wall of the pharynx. The lower airway is measured from the point where the airway is crossed by the mandibular border to the wall of the pharynx [Figure 7].^[5] Neurosensory deficits if any were recorded using standard neurosensory tests.

The selected patients were operated under general anesthesia. A submandibular incision was used to expose the predetermined osteotomy site [Figure 8]. When intraoral technique was employed, an incision was placed on the anterior margin of the ramus, the vertical limb till the distal of the second molar, and the mesial release till the distal of the second premolar. Using a reciprocating saw, the corticotomy was done under copious normal saline irrigation.

The distractor placement was marked by drilling holes. The cuts were deepened till the buccal and the lingual corticotomy was completed with smith spreader, simultaneously taking care of the neurovascular bundle. The prior selected distractor device was then applied on the lateral surface of the mandible and was adapted precisely. The distractor was fixed into the predrilled orientation holes. The distractor was checked before the closure. The flaps were closed using sutures taking care to see that the distractor arm was protruding from the anterior release and resting in the vestibule to facilitate easy activation.

After the required latency time period of 5 days, activation was initiated by 0.5 mm twice daily. Following the latency period of 5days, the distracters were activated with the manufacturer provided activator. The distraction was initiated with 0.5 mm of activation twice a day i.e., 360° clockwise rotation results in 1mm distraction. The patients were discharged after completion of the distraction protocol and were counseled on the maintenance of oral hygiene and diet.

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Figure 1: Preoperative clinical picture



Figure 2: Postoperative clinical picture

The devices were left intraorally for a consolidation period of 90 days, and the bone formation was assessed by the radiographs during the consolidation period. The removal of the devices was done after completion of the consolidation stage. Evaluation of the distraction technique was done on the basis of clinical examination of the patient, distraction device, and the postoperative complications as per when the patient reported for follow-up.

Mean was calculated for the parameters. On comparison, independent *t*-test was used to compare the right and left side measurements. One way ANOVA test was done to assess the significance in comparison of preoperative and postoperative measurements. P < 0.05 is considered as statistically significant and <0.01 is considered as highly significant. P > 0.05 is considered as nonsignificant.

RESULTS

There was overall improvement in esthetics; both soft and hard tissue enhancement was noted. There was considerable improvement in airway. The result of DO was assessed clinically and radiographically between the previously mentioned fixed points for all individuals. The results were tabulated under the following headings.

1. Table 1 – Demographic data and results of the study

- 2. Table 2 Preoperative and postoperative soft tissue clinical dimensions recorded at various intervals
- 3. Table 3 Preoperative and postoperative hard tissue radiographic dimensions recorded at various intervals.

All the patients in the study showed significant improvement in the facial esthetics and improvement in the airway and quality of life. Pain and temporary paresthesia were the common complaints during the distraction. There were two cases, where there was a soft tissue injury due to impingement of the activation arm. In one case, the activation arm was broken during the period of distraction. However, the desirable results overweighed the very few complications.

DISCUSSION

Harmonious and balanced jaws constitute for the functional and esthetic harmony in human organisms. Various craniofacial anomalies involve the lower jaw more commonly than the upper jaw. Numerous surgical techniques have been advocated to correct the mandibular deformities which include mandibular osteotomies with or without the combination of bone grafts. The major hurdle encountered for these surgical procedures is rudimentary bony anatomy, which makes traditional osteotomies difficult to perform. The other hurdle is that the deficiencies with greater magnitude when acutely stretched have more relapse rates and impaired Alwal, et al.: Soft tissue and hard tissue changes and airway improvement with distraction osteogenesis in mandibular micrognathia

Cases	Age/sex	Diagnosis	Osteotomy site	Lengther	ning (mm)	Complications				
				Left	Right					
1	21/female	Mandibular hypoplasia	Bilateral body of the mandible	12	12	Temporary dysesthesia for 1 month Pain during first few days of distraction				
2	17/male	Postankylotic deformity	Bilateral body of the mandible	12	12	Pain at the distraction site during the days of activation				
3	19/male	Postankylotic deformity	Bilateral body of the mandible	15	15	Injury to the lip on both sides due to the impingement of the distraction arm during the distraction period				
4	14/female	Mandibular hypoplasia	Right body and left angle	15	15	Temporary dysesthesia. Breakage of the activating arm of distractor during distraction				
5	14/female	Postankylotic deformity	Bilateral body of the mandible	15	15	Pain at the distraction site during the days of activation and soft tissue injury due to the activation arm during the consolidation period				

Table 1: Patient demographics and results of the study

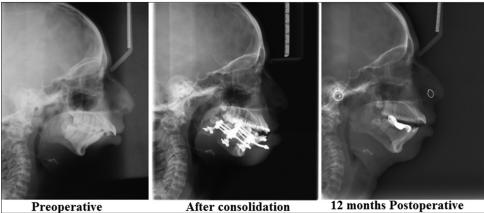
Table 2: Pre- and post-operative soft tissue clinical dimensions recorded

Case	Advancement		vancement Medial canthus to buccal commissure length (mm)				Lateral canthus to buccal commissure length (mm)				Lateral canthus to external acoustic meatus length (mm)				Buccal commissure to external acoustic meatus length (mm)			
	LT	RT	Preoperative	3 m	6 m	12 m	Preoperative	3 m	6 m	12 m	Preoperative	3 m	6 m	12 m	Preoperative	3 m	6 m	12 m
1	12	12	60	66	66	65.5	69	75	74	74	66	66	67	67	87	96.5	96	95
2	12	12	63	69	69	68.5	67	74	73	73.5	71	71	71	70	89	95	95	94
3	15	15	64	71	70	69.5	75.5	82.5	82	81.5	74	75	74	74	92	99	98.5	98
4	14	12	58.5	65	64.5	64	61.5	67	66.5	66	64	65	65	65	89.5	95	94	94
5	14	14	54.5	64.5	63.5	63	58	64.5	64	63.5	58	58	58	58	83	92	92	91
Mean	13.4	13.0	60	67.1	66.6	66.1	66.2	72.6	72	71.5	66.6	67	67	66.8	88	95.5	95.1	94.4
Ρ			0.006**,b			0.469 ^b			0.999 ^b				0.001**, ^b					

*P value of independent t-test, *P value of one-way ANOVA test. *Statistically significant (P<0.05), **Highly significant (P<0.01). LT: Left, RT: Right



Figure 3: Orthopantomographs



12 months Postoperative

Figure 4: Lateral cephalograms

nerve function. These reasons often necessitate multiple surgeries and the use of bone grafts. All these orthognathic surgeries were aimed at correcting the skeletal deformity,

but the soft tissues were just spoken.^[6] DO is a technique for creation of new bone during significant lengthening of the mandible without the demand for bone grafting

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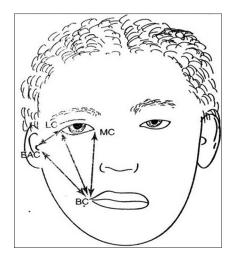


Figure 5: Soft tissue landmarks

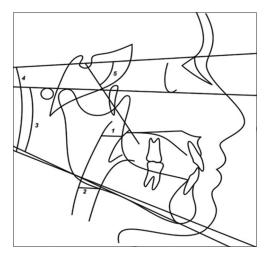


Figure 7: Airway analysis

and associated donor site morbidity. This has become increasingly popular after McCarthy showed the feasibility of lengthening hypoplastic mandibles. Since then, the method of DO to correct mandibular hypoplasia has been extensively used.^[7] Distraction forces applied to bone create a stress in the surrounding tissues, resulting in a sequence of adaptive changes in the soft tissues allowing larger skeletal movements while minimizing the relapse.^[8] In this study, an effort was made to assess the efficiency of DO in lengthening the mandible and associated soft tissue alterations.

The technique of DO as in our study can be applied in growing age group. Tehranchi and Behnia^[9] stated that the reconstruction of severe hypoplastic mandibles in young children is generally addressed with a costochondral bone graft. The capriciousness of its growth and other complications makes the usage of DO to be preferred over costochondral graft reconstruction. An important extra benefit of the gradual distraction is that it not only lengthens bony skeleton but also the associated soft tissues, such as

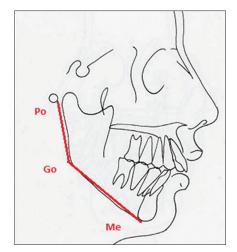


Figure 6: Hard tissue landmarks

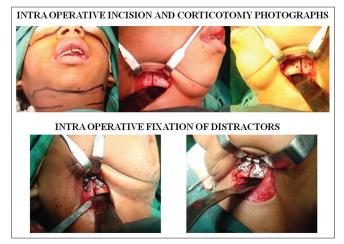


Figure 8: Intraoperative pictures

the muscle, subcutaneous tissue, and skin. Because of the expansion of the associated soft tissues, there is a resulting multidirectional expansion of the skeletal and soft tissue envelope.^[10] In our study of 5 cases, patient's mean age was 17 years. There was a good improvement in the soft tissue landmarks coinciding with the hard tissue changes.

In general, two types of devices have been used for craniofacial osteodistraction: external and internal. The earliest devices used for DO were of the external type, which were primarily applied for larger advancements. Although the external devices are proven to be more advantageous, the hardware and external scars made them uncomfortable and unacceptable. Maull^[11] reviewed various devices for DO in the craniofacial complex. The distraction devices are categorized based on whether they are internal or external, tooth borne, or bone borne. The devices may be unidirectional, bidirectional, or multiplanar. The distraction devices are employed to lengthen the mandibular ramus, mandibular body or to widen the mandible or to augment the alveolar ridge.

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Case	Advancement (mm)		Vertical length of ramus (mm)				Horizontal body length (mm)				Upper airw	Lower airway length (mm)						
	LT	RT	Preoperative	3 m	6 m	12 m	Preoperative	3 m	6 m	12 m	Preoperative	3 m	6 m	12 m	Preoperative	3 m	6 m	12 m
1	12	12	43	46	46	45.5	54	65	64	63	6	16	16	15	4	10	10	10
2	12	12	36	43	42	42	35	43	43	42	3	8	8	7	2	7	7	7
3	15	15	51.5	58	57.5	57	49	58	57	57	8	14	14	12	7	10	9	9
4	14	12	57	66	66.5	66	68	74	74	73	6	10	10	9	5	9	9	8
5	14	14	55.5	62	62	61	55	63	62	62	6	14	13	13	6	12	12	12
Mean	13.4	13.0	48.6	55	55	54.5	52.2	60.6	60	59.4	5.8	12.4	12.2	11.2	4.8	9.6	9.4	9.2
Ρ	0.698ª		698ª 0.702 ^b			0.633 ^b			0.007** ^{,b}				0.002** ^{,b}					

Table 3: Pr	e- and	post-operative	hard tissue	radiographic	dimensions	recorded

^a*P* value of independent *t*-test, ^b*P* value of one-way ANOVA test, *Statistically significant (*P*<0.05), **Highly significant (*P*<0.01). LT: Left, RT: Right, 3m: 3-month postoperative, 6m: 6-month postoperative, 12 m: 12-month postoperative

The advantages of mandibular DO using intraoral devices identified by Primrose *et al.*^[12] are as follows:

- 1. Absence of facial scars
- 2. Better patient acceptance due to small and inconspicuous nature of the device
- Design of devices based on anatomic location (corpus or ramus)
- 4. Improvement of osteodistraction techniques
- 5. Design of devices based on clinical application (lengthening or widening).

In our study, internal monodirectional distraction devices made of stainless steel were used and they have shown excellent patient acceptance. In a survey done by Ali *et al.*,^[13] in 2009, the histologic and the cellular events were analyzed. They concluded that DO produced bone through intramembranous and endochondral bone formation along the vector of distraction. The phenomenon of Callotosis is a gradual stretching of the reparative callus forming around bone segments. In our study, we found the similar result; the radiolucency which was present at the distraction site immediately after distraction had gradually underwent ossification resulting in the formation of radio-opacity at the distraction gap during the period of consolidation.

Every orthognathic procedure is tied in with a certain definite quantity of soft tissue alterations.^[14,15] Sparse data is available in literature in relation to the soft tissue changes associated with DO. The clinical analysis was done by Mikhal I. Samchukov *et al.*,^[3] using the distances between fixed reference points on the face. Linear measurements between these fixed reference points were counted at various pre- and post-operative time intervals to quantify soft tissue change. Soft tissue changes during DO were studied by Apaydin *et al.*,^[16] with histomorphometric analyses. The authors concluded that the number of muscle fibers and nuclei on the distraction side increased proportionally with the distraction period thus increasing the bulk of the soft tissue. The clinical observations in our study showed that

there was a rapid descent of the BC and an increased distance between the BC and the canthi. There is statistically highly significant difference present between the preoperative and postoperative measurements of medial canthus to BC length with gradual increase in the measurement at 6 months' interval with P = 0.006. There is a statistically high significant difference between the preoperative and postoperative measurements of buccal commissure (BC) (figure 5) to external acoustic meatus (EAM). There was greater statistical significance by end of 3rd month (P = 0.001).

There was a markable improvement in the facial balance in all the patients. It was also observed that there was extra increase in the volume of the cheek, which is probably related to the improved muscular activity and increase in the length of the underlying bone. The similar finding was observed by Trahar *et al.*^[17]

There is recognizable increase in the vertical and horizontal dimensions in the mandibular body length in the study. The hard tissue changes achieved by cephalometric analysis in the horizontal plane have shown lengthening of the mandible along with additional significant increase in the height of the vertical ramus when an oblique osteotomy was made and an oblique vector was used for distraction. Rubio-Bueno et al.^[18] noticed similar finding in their report. Fullness of the cheek as seen in the clinical evaluation can be correlated to an increase in the height of the vertical ramus leading to the descent of the Gonion. The increase in the distance between BC to the canthi can be correlated to the increase in the corpus length. This determination is similar to the findings in the study by Fu et al.^[19] In our study, we sought to evaluate soft and hard tissue changes using clinical and cephalometric analysis which has respectively produced encouraging results in both soft and hard tissues. There was an overall growth in the bone and soft tissues clinically and radiographically.

Several other authors compared the stability of mandibular DO with that of the bilateral sagittal

split osteotomy (BSSO). They concluded that similar relapse rates between DO and BSSO were observed in mild to moderate advancements of 6-10 mm, but the neurosensory disturbances and condyle resorption were more in the later technique. Schreuder et al.^[20] found that in the patients with low mandibular plane angle and in patients with advancement of >7 mm, DO is preferred over BSSO, and in the patients with advancement <7 mm, the results for the both operative procedures were comparable. van Strijen^[21] et al. evaluated the stability of DO in mandibular advancement and concluded that it is a safe procedure in the patients with low mandibular plane angle. In our study, the constancy of the progression of the mandible was compared till 12-month postdistraction period. The average advancement in our survey was 12.6 mm. The advancements showed greater stability with only 3% relapse in the mandibular corpus length because the overlying soft tissues are stretched simultaneously thus minimizing the relapse.

In summation, to the satisfactory and significant results, we took in our share of complications. All the patients had pain during activation of the distractor. This feature is anticipated as the forces exerted at the osteotomy site. One of our patients experienced hypoesthesia of the inferior alveolar nerve during distraction which resolved in 2-month postoperatively. This can be attributed to the gradual stretch of the nerve during advancements which can lead to an acute injury of the nerve. Van Strijen et al.^[22] in their study on the complications of bilateral mandibular DO, stated that the nerve gradually adapts to the new length and provides the patient with better sensory recovery and function. The authors experienced complications in 40% of the cases. The complications in the study were technique or device related. There was no infection or serious complications that required rehospitalization. The neurosensory deficit was temporary which regained sensation by the end of the study. There was no permanent neurosensory deficit. Hence the writers conclude that DO could be reckoned as a safe and predictable procedure for lengthening the mandible, with a low incidence of major complication. The writers concluded that DO can be reckoned a safe and predictable procedure for lengthening the mandible, with low incidence of major complication. In this study, we experienced similar complication of breakage of the distraction device at the activation arm during the dynamic period of distraction in one case. There was a transient neurosensory deficit in two cases, and in two patients, there was impingement of the activator arm into the lower lip. Pain was experienced by the patients during activation which was expected. Breuning et al.^[23] conducted a study to quantify the overbite that occurred during the advancement by DO.

The authors suggest that opening of the bite is expected complication of DO. One patient has made an open bite, which was further adjusted by postsurgical orthodontics.

Bouchard *et al.*^[24] studied the use of DO in the treatment of obstructive sleep apnea (OSA). The authors summarized that DO of the facial skeleton is an alternative choice to standard orthognathic surgery for selected patients with OSA, and the technique of DO allows large advancements without the demand for bone grafting, less danger of relapse, and less chance of inferior alveolar nerve injury. Our work resulted in an excellent improvement of both upper and lower airways, thereby proving DO as an excellent method for the treatment of OSA. The patients were relieved of snoring and thereby improved the quality of life.

Apart from the device-related complications, the intubation was difficult in the patients with severe micrognathia. The reason for the difficult intubation was retruded position of the mandible and the associated soft tissue structures. The methods for intubation in such patients are laryngeal mask airway, fiberoptic-guided intubation, retrograde intubation, and blind nasal intubation. When all the above methods fail, tracheostomy is the option. An important disadvantage which was observed in the study with internal distractors is the need for second surgery for the removal of the distractors. Although the outcome was more than satisfactory, a longitudinal prospective study with larger sample size will provide more coherent and sound information for the technique.

CONCLUSION

The surgical technique of DO is similar to that of conventional orthognathic surgery but with an advantage of slow bone movement and concomitant expansion of the soft tissue envelope. With the development of intraoral devices, it has allowed better patient acceptance of this procedure. When using a novel technique to clinical situations where other techniques are available, the risks and benefits of the new technique must be compared with commonly used surgical procedures. In our study, DO was used in 5 patients with severe mandibular deficiency. In all the patients, we obtained correction of deformity and improvement of the airway with minimum complications. Despite fewer complications, DO is a good surgical modality for the correction of extreme micrognathia. It is comparatively dependable and effective procedure that can be done even during the growth phase. In spite of the technique having been practiced for over 20 years, no concrete long-term solutions are put down. It requires including adequate numbers in samples, possibly multicenter study, treated in a standardized manner and documented and evaluated properly to have meaningful conclusions.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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